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Published in:

Journal of Cleft Lip Palate and Craniofacial Anomalies

DOI:

[10.4103/2348-2125.187510](https://doi.org/10.4103/2348-2125.187510)

Publication date:

2016

Licence:

No Licence / Unknown

Document Version

Peer reviewed version

[Link to publication in Discovery Research Portal](#)

Citation for published version (APA):

Qureshi, N., McIntyre, G., & Mossey, P. (2016). Does relative maxillary arch constriction worsen with growth in patients with Cleft lip and palate? A pilot study. *Journal of Cleft Lip Palate and Craniofacial Anomalies*, 3(2), 77-82. <https://doi.org/10.4103/2348-2125.187510>

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Original article

Article title

Does relative maxillary arch constriction worsen with growth in patients with Cleft lip and palate? A pilot study

Running title

Maxillary growth in CLP

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Article structure

Total pages: 12

Photographs: 6

Word counts: abstract = 322, main text = 2273, introduction = 697, discussion = 1213

Authorship statements

The authors declare that there are no conflicts of interest / financial relationships with any product or service described in the manuscript

All authors were involved in conception and study design, experimental procedures, drafting, writing and revising the manuscript. The final version of the manuscript has been read and approved by all the authors and that each author believes the manuscript represents honest work.

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ABSTRACT

Objective: To evaluate relative maxillary arch constriction in patients with unilateral cleft lip and palate (UCLP) and isolated cleft palate (CP) during growth and the reproducibility of a commercial 3D small object scanner.

Design & Setting: Retrospective longitudinal study, University Dental Hospital.

Materials and methods: Plaster models at age 5, 10 and 15 years of ten patients with UCLP and CP were scanned using a commercial 3D small object scanner (www.NextEngine.com). Three observers scored the plaster and digital models using the 5 year old/GOSLON and Modified Huddart Bodenham (MHB) indices and again 3 weeks later. Two-way ANOVA ($P < 0.05$) calculated the significance of the differences between age and group (UCLP and CP). Weighted Kappa and Kendall's coefficient values determined agreement within and between the observers.

Results: There were no statistically significant changes in the occlusal index scores with age ($P > 0.05$). For plaster models, intra- and inter-observer reproducibility were good to very good (0.74–0.84 and 0.84–0.86) using 5 year old/GOSLON indices compared with good to very good (0.72–0.85) and very good (0.84–0.86) for the MHB index. For digital models, there was moderate to good intra-observer (0.46–0.79) and inter-observer (0.44–0.67) reproducibility for the 5 year old/GOSLON indices compared with good intra- (0.66–0.75) and inter-observer reproducibility (0.68–0.71) with the MHB index.

Conclusions: There was no progressive worsening of relative maxillary arch constriction with growth in UCLP and CP. The reproducibility of the 5 year old/GOSLON and MHB indices was acceptable with digital models produced using the commercial 3D small object scanner but good to very good for plaster models. Longitudinal evaluation of maxillary growth in patients with repaired orofacial clefts using these methods is

both informative for the care of the individual patient and an important step in refining surgical protocols in countries such as India.

Does relative maxillary arch worsen with growth in patients with Cleft lip and palate? A pilot study

Introduction

Orofacial clefts are a heterogeneous group of congenital disorders characterised by the presence of fissures on the lip and/or the palate.^[1] In vast countries such as India where there is a dynamic genetic pool with wide cultural and ethnic variations, the prevalence of cleft lip and palate varies from state to state. There is no comprehensive registry and reporting of new cases of orofacial clefts is inadequate due to lack of the infrastructure in rural areas and disparity of the healthcare facilities between rural and urban areas.

The precise aetiology of CLP remains to be fully determined with progress having been made in identifying the environmental factors and genes involved in syndromic CLP. The current evidence suggests clefting is multifactorial in nature with a genetic predisposition and contributing environmental factors.^[2] The primary surgical repair of the lip and palate is one of the earliest and most significant interventions that is carried out for the management of patients with CLP. There are multiple treatment protocols used by cleft teams worldwide and there is no consensus about the optimal timing and techniques for surgical repair. The UK clinical standards advisory group (CSAG) identified that many cleft centres had adopted a variety of differing surgical protocols.^[3] Evidence-based practice should answer uncertainties for the treatment of patients with clefts; however high quality evidence (systematic reviews and randomised controlled trials) of cleft lip and palate care is scarce.^[4] Multicentre collaboration among treatment providers has the potential to reduce the variability of treatment protocols and ensure that patients with clefts receive evidence-based clinical care.^[5] In recent years orthodontists have been instrumental in large scale multicentre randomised clinical trials for the assessment of CLP surgical outcomes.^[6] Poorly performed surgery carries a high risk of interference with facial growth, dental development and speech with maxillary retrusion being a common

long-term complication^[7] characterised by collapse of the maxillary arch with resultant anterior and/or posterior crossbites. Reiser et al^[8] evaluated the changes in cleft size and maxillary arch dimensions and related these changes to the surgical interventions in UCLP and CP. The patients with UCLP had wider maxillary arch dimensions than the patients with CP during their first year of life, when only lip closure was undertaken. After the closure of hard palate, the transverse growth reduced in patients with UCLP. At age 5, the patients with UCLP had similar maxillary arch dimensions compared to patients with isolated CP.

The 5 year old, GOSLON yardstick and the modified Huddart/Bodenham index (MHB) are routinely used to evaluate the outcomes of surgery. The GOSLON yardstick is the most commonly used index for the measurement of surgical outcome worldwide but is only suitable for the late mixed dentition.^[9] The 5 year old index was developed to allow surgical results to be assessed in younger patient.^[10] However, both are 5-point categorical indices whilst, the MHB index is an ordinal scale index that can be used at any age. As a result, the MHB index allows more precise occlusal differences to be detected, enabling the determination of more subtle differences between groups.^[11]

Various methods have been used to measure relative maxillary arch constriction including dental models, photographs and cephalometric radiographs. Plaster models have a number of drawbacks including the burden of storage, fragility and loss. Various methods have been used to archive plaster models into three-dimensional digital models including stereophotogrammetry, holography and laser scanning. The latter have been validated for use in cleft care^[12-17] with digital study models increasingly being used for orthodontic diagnosis and record keeping.^[18] However the production of digital models requires the use of specific scanning equipment, which is expensive. The quality of 3D digital dental models produced using a low cost commercially available 3D small object scanner for use in cleft care in countries such as India where cleft care resources are unequally spread and where access to multidisciplinary cleft care in large areas of the country is limited has not been tested to date.^[19]

The null hypotheses were:

1. Relative maxillary arch constriction in patients with UCLP and CP does not deteriorate progressively with growth.
2. The reproducibility and reliability of the 5 year old/GOSLON and MHB indices are similar when assessed on plaster and digital models.

Materials and method:

This was a retrospective study of patients with non-syndromic CLP (five UCLP and five CP) who were randomly selected from the Cleft Care Scotland archive. All had undergone surgical repair and orthodontic treatment. Caldicott Guardian approval in relation to consent was provided by NHS Tayside, access to data was granted by Cleft Care Scotland and all data were handled in accordance with the Helsinki Declaration of 1975 and subsequent revisions. The plaster models recorded at age 5, 10 and 15 years were scanned in occlusion using a commercially available desktop 3D Laser scanner (www.NextEngine.com) at 127 microns accuracy. The two scanned families were superimposed and the resultant mesh points were converted into a final scanned model by ScanStudio HD software (Figures 1, 2). The scanned models were saved on a password protected laptop (www.acer.co.uk). The plaster and digital models for each patient were then scored using the 5 Year old/GOSLON and modified Huddart/Bodenham indices by three observers on two occasions, 3 weeks apart. The data were entered into a spreadsheet (Microsoft Excel, Redmond, California) for statistical analysis (www.Rstudio.com). Two-way ANOVA was used to assess the differences in relative maxillary arch constriction with growth between the UCLP and CP groups. Weighted Kappa and Kendall's correlation coefficient were calculated to determine intra- and inter-observer reproducibility for the plaster and digital models with the values categorised according to the Altman method.^[20]

Results

Although the 5 year old/GOSLON/MHB scores for UCLP and the MHB score for CP improved from age 5 to 15, the 5 year old/GOSLON score for CP improved to a lesser degree from age 5 to 15 (Figures 3, 4). These changes were not statistically significant ($P > 0.05$) (Table 1).

Therefore the first null hypothesis was accepted.

For digital models, intra- and inter-observer reproducibility was moderate to good for the 5 year old/GOSLON indices compared with good intra- and inter-observer reproducibility with the MHB index. For plaster models, intra- and inter-observer reproducibility were good to very good using 5 year old/GOSLON indices but were good to very good and very good, respectively for the MHB index (Tables 2 and 3) The second null hypothesis was therefore rejected.

Discussion

It was found that relative maxillary arch constriction did not deteriorate progressively with growth, but this was greater in patients with surgically treated UCLP than in isolated CP. As a result, growth and relative maxillary arch constriction are independent of each other. Furthermore, the reproducibility of the indices was generally better with plaster models than digital models. The MHB index performed better on digital models when compared to the 5 year old/GOSLON index indicated its superiority over categorical indices for the assessment of cleft care outcomes.^[21] It was noted that the surgically treated 5 year old patients had a relatively constricted maxilla in relation to the mandible in both groups (UCLP and CP) with the level of constriction showing a decreasing trend in all patients at age 10. The decrease in relative maxillary arch constriction continued to age 15 as confirmed by the lower ranks of GOSLON scores and the near zero or positive scores for the MHB index data when compared to the corresponding scores at age 5 and age 10. This may have been due to orthodontic treatment having increased maxillary arch width by age 15 even though the growth of the maxilla and mandible is not complete at this age.

Relative maxillary arch constriction is one of the most common long-term complications of cleft surgery, with the aetiology being manifold. Relative maxillary arch constriction results from scarring due to primary surgery^[22] and an intrinsic growth deficiency in patients with CLP.^[23]

The present study identified non-significant growth differences between two cleft types regarding relative maxillary arch constriction, indicating that there is no specific trend of maxillary growth deterioration with time. Nevertheless, subjects with UCLP had a greater degree of relative maxillary arch constriction

compared to patients with CP. The likely causes of this are manifold and include the fact that in UCLP compared to CP, the original tissue defect is larger and affects the alveolus, and whilst CP generally requires a single operation, UCLP requires a minimum of three operations. The resultant restriction on facial growth from these surgical interventions restricts facial growth to a greater extent in patients with UCLP than in patients with isolated CP. It is interesting to note that despite the cleft size and morphology of the maxilla in infants with UCLP being different to that in infants with CP, the primary cleft repair and subsequent treatment protocols for both cleft sub-phenotypes are similar but resulting in different treatment outcomes.^[8]

Previous researchers have also identified abnormal muscle forces in the repaired lip of patients with UCLP when analysed electromyographically.^[24] The interaction of disordered muscle forces on the maxilla and dentition in UCLP results in further occlusal and skeletal differences when compared to CP. The disturbance in lip musculature in the UCLP group could affect the normal growth of the maxillary complex, thereby producing greater relative maxillary arch constriction in this group when compared to patients with CP. Although the reduction of maxillary width is associated with a disturbance in the palatal sutural system in patients with cleft palate,^[25] the combination of scarring caused by surgical repair of both the cleft lip and cleft palate results in significant growth restraint and maxillary hypoplasia in UCLP.^[9] Vandas^[26] found that 3 year old patients with treated UCLP had a smaller maxilla and a normally sized mandible and patients with treated CP had a smaller maxilla and mandible (by a similar magnitude) when both were compared with children without clefts. Our results are in keeping with these findings where relative maxillary arch constriction was greater in patients with UCLP compared to patients with CP.

Recently, various 3D scanners have been introduced to produce digital models. The NextEngine desktop laser scanner was used to assess if a cost-effective small object scanner would be adequate to produce digital models for the assessment of dental arch relationships. Whilst no study has used it for plaster models of patients with CLP it has been used directly on patients' faces^[27] and this study supports the use of the NextEngine scanner for the production of digital models. Weighted Kappa was used with the categorical data (5 year old and GOSLON indices) and Kendall's correlation coefficient was used for the ordinal data (MHB) to determine intra- and inter-observer reproducibility. The former takes into account the magnitude

of the difference between scores (i.e., scores of 2 and 3 are relatively close, but 2 and 5 are a long way apart). The reproducibility of the data was generally greater with plaster models compared to digital models, which was nevertheless acceptable. The possible reasons for the difference are manifold, the resolution of the scanner used in this study had an accuracy of 127 microns compared to other scanners with a higher resolution (10 microns), where the quality of the digital models is greatly influenced by scanner resolution. Only one occlusal position exists for the digital models compared to plaster models where the occlusion can be subtly altered during scoring. Furthermore, when using digital models, planes and angles are more difficult to assess than using plaster models making the scoring potentially less reliable.^[18] As we found good or very good reproducibility with digital models, a commercial small object scanner can therefore be used for the evaluation of cleft care outcomes instead of plaster models in remote areas of countries such as India where there is no access to a proprietary laboratory based model scanner.

Due to reduced accessibility or poor quality of cleft care in developing countries, non-government organisations such as Smile train and Transforming faces worldwide concentrate on the provision of primary surgical procedures in the countries like India.^[28] Cleft care provided by these humanitarian organisations should be expanded to provide pre-operative evaluation and post-operative care. Making use of innovative model scanning technology and wireless 3G connections available worldwide would allow cleft care outcomes in countries such as India to be scored remotely in accordance with increasing research initiatives.^[19]

Although the study is significantly underpowered, this paper describes the methodology that could be applied to future studies that might be carried out in order to cast light on the important question of how predictive the dental arch dimensions of patients with repaired orofacial clefts at the age of 5 are for arch parameters at the pre-adolescent age of 10, and after the pubertal growth spurt at the age of 15, as there are significant changes in dental development and facial growth over this period.

Further research is required to investigate maxillary growth in UCLP and CP along with other cleft sub-phenotypes including bilateral cleft lip and palate (BCLP), cleft lip and soft palate clefts to determine the nature of any differences in the pattern of maxillary growth and how this relates to surgical protocols.

Conclusions

1. There was no progressive worsening of relative maxillary arch constriction with growth between ages 5 and 15 years in patients with UCLP and CP.
2. The reproducibility of the 5 year old/GOSLON and MHB indices was acceptable with digital models produced using the commercial 3D small object scanner but good to very good when using plaster models.
3. Longitudinal evaluation of maxillary growth in patients with repaired orofacial clefts using these methods is both informative for the care of the individual patient and an important step in refining surgical protocols in countries such as India.

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Figures

Figure 1: Digital models of a patient with CP: A: 5 years old, B: 10 years old, C: 15 years old

Figure 2: Digital models of a patient with UCLP A: 5 years old, B: 10 years old, C: 15 years old

Figure 3: 5 year old/GOSLON scores for plaster models

Figure 4: MHB scores for plaster models

Tables

Table1: Differences between occlusal scoring systems with growth for UCLP and CP groups

Table 2: Intraobserver reproducibility for 5 year old/GOSLON and MHB Scores with plaster and digital models

Table 3: Interobserver reproducibility for 5 year old/GOSLON and MHB Scores with plaster and digital models